



Endoscopic findings of esophageal submucosal gland duct adenoma

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A 78-year-old woman with chronic GERD presented to the primary care office for evaluation of dyspepsia and dysphagia. Upper endoscopy revealed a partially obstructing submucosal tumor 15 cm from the incisors. EUS revealed a 2-cm submucosal mass, and the patient was referred to our institution for therapy.

A smooth, 2-cm lesion (spanning from 15 to 17 cm from the incisors) that was obstructing one-third of the lumen was inspected on white-light examination (Fig. 1). On EUS, the mass was localized to the deep mucosa and

submucosa, had well-defined borders, and had a macrocystic appearance (Fig. 2). The cysts were prominently sized, and no vascular flow was noted on color Doppler within the cystic lesions to suggest the presence of varices. Other submucosal tumors often found in the esophagus were considered, such as fibrovascular polyp, duplication cyst, and granular cell tumor; however, the location and EUS appearance were inconsistent with these etiologies. Given that the patient was symptomatic and the mass did not involve the muscularis propria, endoscopic submucosal dissection (ESD) was performed.

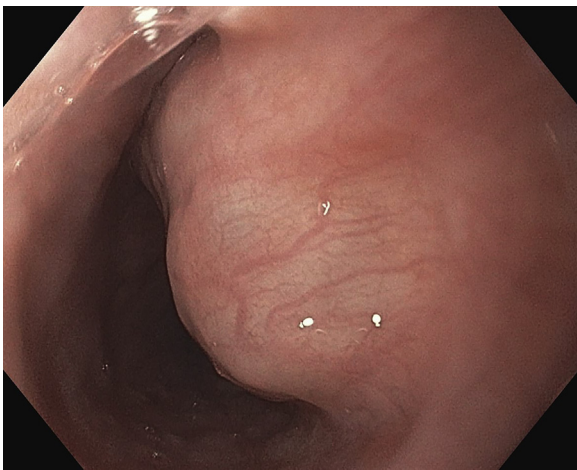


Figure 1. White-light examination revealing a partially obstructing submucosal mass in the proximal esophagus. The normal mucosa gives the appearance of a submucosal mass.

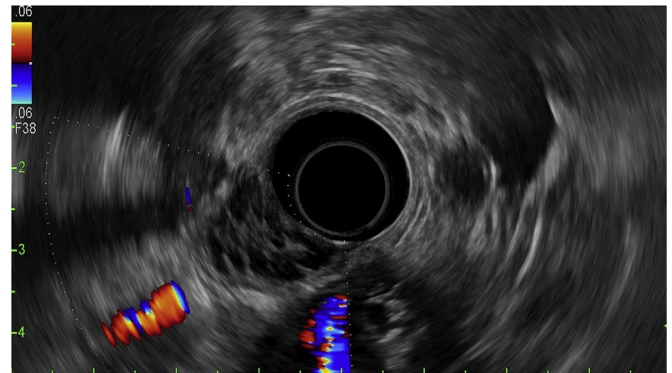


Figure 2. EUS view showing a well-circumscribed, multicystic, submucosal mass at the 8 o'clock position. The cystic lesions are prominent and show no signs of vascular flow on color Doppler.

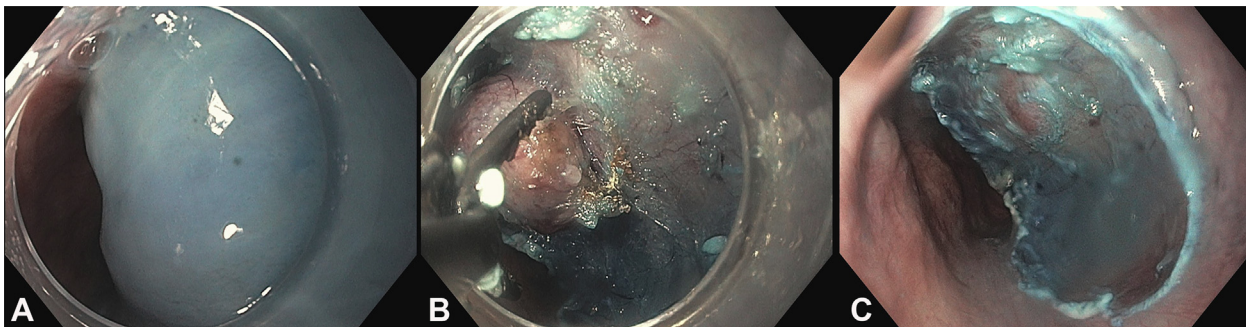


Figure 3. Endoscopic submucosal dissection. **A**, Submucosal injection. **B**, Endoscopic submucosal dissection with an articulating knife, arms open. **C**, Resection bed after ESD.

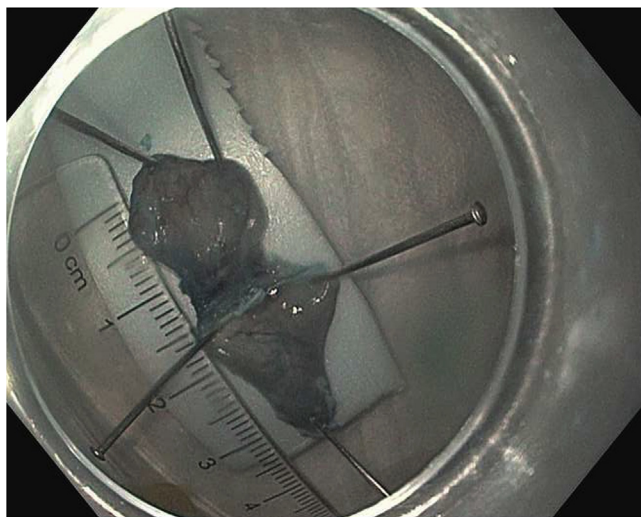


Figure 4. En bloc gross specimen.

We performed ESD with an articulating endoscopic knife that permits a grasp, pull, and then cutting technique to allow precise application of electro-surgical energy and coagulation during dissection. The tumor was lifted with a solution of 15% hydroxymethylcellulose, methylene blue, and 1:200,000 epinephrine injection solution. Circumferential markings were initially made; then, a circumferential incision was completed. When we arrived toward the deeper tumor margins, the articulating knife was then used in a novel fashion to blunt-dissect the base of the lesion from the muscularis propria, without need for a cutting current (Fig. 3; Video 1, available online at www.VideoGIE.org). An en bloc specimen was resected for histopathologic analysis (Fig. 4). Histologic examination showed multiple lobulated cystic proliferations of 2 layers of benign epithelium within the submucosa, consistent with esophageal submucosal gland duct adenoma ESGDA (Fig. 5).

ESGDA is a rare tumor arising from submucosal gland duct clusters in the esophagus.¹ Patients may present with dysphagia or dyspepsia; commonly, they may be asymptomatic.¹ Historically these have been mostly found in the middistal esophagus, and EUS demonstrates a heterogenous submucosal tumor with many small cystic areas involving the second and third sonographic layers.²⁻⁵ Histologic examination is needed to confirm a diagnosis of ESGDA and will show 2 or more layers of epithelial cells and papillary folds, infrequent mitosis, mild to moderate atypia, and lymphocytic infiltration.¹

Our patient showed important endoscopic variations from what has been observed in the literature on ESGDA (Table 1). Although clusters of esophageal submucosal glands can be found at both the proximal and the distal esophagus,⁶ almost all cases of ESGDA previously reported in the literature were located in the distal esophagus.²⁻⁵ The present case demonstrates that these lesions can also occur in the proximal esophagus, which is biologically plausible, given the known distribution of submucosal gland ducts.²⁻⁶ The EUS findings were also different in that larger multicystic structures were visualized than the expected, small cystic lesions previously reported.^{2,5} This may represent a higher degree of accumulated secretions and cellular debris within the ducts, relating to the larger tumor size in our case compared with others reported (Table 1).

Given that ESGDA is very rarely encountered, it is important to recognize that our understanding of these neoplasms is limited. Reports that describe esophageal cancers arising from submucosal glands suggest a potential biologic pathway for ESGDA to progress to malignancy; thus, resection of these tumors is advised.^{1,2,7-10} ESD not only confirms the diagnosis of ESGDA but also permits complete en bloc resection of these neoplasms, which is the preferred therapeutic management strategy.^{1,2,7-10}

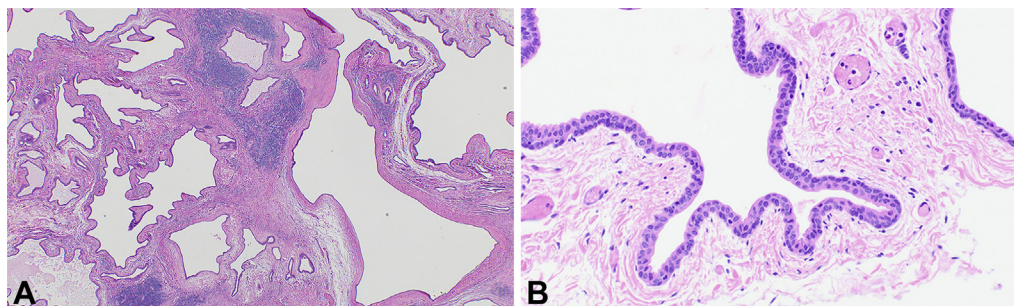


Figure 5. Histologic review of resected specimen. **A**, Multiple lobulated cystic epithelial proliferations with occasional lymphocytic infiltrates within the esophageal submucosa (H&E, orig. mag. $\times 20$). **B**, On high magnification, the cysts consist of multiple (usually 2) layers of epithelial cells/myoepithelial cells with mild cytologic atypia and rare to absent mitosis (H&E, orig. mag. $\times 200$).

TABLE 1. Literature review on endoscopic, sonographic, and histologic characteristics of ESGDA

Study	Location	Size and character	Mucosa	EUS results	Histologic diagnosis
Shibata et al, 2017 ²	Distal esophagus	5-mm mass	Normal	Heterogenous, hypoechoic submucosal mass with tiny anechoic areas	Dilated ducts with 2 layers of proliferating epithelial cells in the submucosa
Hayashi et al, 2004 ³	Mid-esophagus	10-mm mass	Normal	Submucosal mass, not further characterized	Cysts with double-layered epithelium located in the submucosa
Chinen et al, 2004 ⁴	Distal esophagus	6-mm mass	Normal	Heterogeneous cystic lesion in the submucosa (third layer)	Multiple cysts with rare mitotic figures and final diagnosis of ESGDA
Agawa et al, 2003 ⁵	Distal esophagus	15-mm mass	Normal	Well circumscribed, heterogeneous, hypoechoic tumor, at second and third layers, with several small cystic lesions	Dilated gland ducts with 2 layers of epithelial cells in the submucosa

ESGDA, Esophageal submucosal gland duct adenoma.

DISCLOSURE

All authors disclosed no financial relationships relevant to this publication.

Abbreviations: ESD, endoscopic submucosal dissection; ESGDA, esophageal submucosal gland duct adenoma; EUS, endoscopic submucosal dissection.

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